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Case Report

Full Mouth Rehabilitation in a Rare Case of Situs Inversus - A Case Report

Dr. Madhusudhan KS[1*], Dr. Bhargavi R. Vojjala[2], Dr. Priya Subramaniam[3]

[1]Associate Professor, Department of Pediatric and Preventive Dentistry, The Oxford Dental College and Hospital,10th Milestone, Hosur Road, Bommanahalli, Bengaluru, Karnataka 560068, India; ORCID: 0000-0002-0658-7581
[2]Postgraduate, Department of Pediatric and Preventive Dentistry, The Oxford Dental College and Hospital. 10th Milestone, Hosur Road, Bommanahalli, Bengaluru, Karnataka-560068, India; ORCID: 0000-0003-0877-8397
[3]Professor and Head of the Department, Department of Pediatric and Preventive Dentistry, The Oxford Dental College and Hospital. 10th Milestone, Hosur Road, Bommanahalli, Bengaluru, Karnataka-560068, India; ORCID: 0000-0002-2935-6524

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Abstract: Situs inversus totalis (SIT) has been reported as a rare entity. It may elude routine patient evaluation, detailed knowledge about the normal anatomical structure and its variations must be known for daily clinical practice and even more critical in an emergency. SIT is inherited as an autosomal recessive pattern and is associated with multiple gene mutations. Situs inversus totalis is a condition that is characterized by abnormal positioning of the heart and other internal organs. It may or may not be associated with other congenital defects. Diagnosis of situs inversus totalis is an important in a patient who does not have any congenital anomalies, which may indicate a normal life expectancy, the patient'spresentation of common ailments may become difficult to diagnose due to mirrored anatomy. Here we report the full mouth rehabilitation in a case of situs inversus totalis.

Keywords: Congential, dextrocardia, situs inversus.

INTRODUCTION

Dextrocardia is an abnormal positioning of the heart in the right side of the chest instead of the left [1]. A condition in which dextrocardia is associated with a reversal in the position of other organs is known as situs inversus totalis. Situs inversus totalis (SIT) is defined as a mirror image reversal of all the asymmetric structures of the thorax and abdomen [2].

Marco Severino in 1643 was the first to describe this condition and it is an autosomal recessive genetic condition. Situs inversus totalis has only occurred in a few cases and its incidence is about 1:10,000 live people [2-4].

Generally, many individuals with SIT are unaware of their unusual anatomy. Situs inversus totalis is often diagnosed while medically assessing other conditions [5]. Diagnostic modalities include chest radiograph, electrocardiogram, echocardiography, and magnetic resonance imaging [6].

Here we present the case of an 8-year-old Indian boy requiring full mouth rehabilitation and diagnosed with situs inversus totalis. Treatment included extractions, pulp therapies, and placement of removable functional space maintainers.

CASE PRESENTATION

Patient Information

An 8-year-old Indian boy was reported to the department with a complaint of retained upper front teeth. Parents also complained that several of the child's teeth had blackish discoloration, were chipped or broken, and the child could not chew properly. On Medical history, parents revealed that the patient was born with situs inversus. The delivery was reported to be normal and there was no history of any illness, trauma, or drug use during pregnancy. The marriage was also not consanguineous. The condition was diagnosed when the patient was 4 months old. At that time, a chest x-ray was taken for treatment of cold and it was discovered that the child had situs inversus. However, the patient did not have any other congenital abnormality. There was no other family history of any congenital anomalies.

***Corresponding Author:** Madhusudhan KS
Associate Professor, Department of Pediatric and Preventive Dentistry, The Oxford Dental College and Hospital,10th Milestone, Hosur Road, Bommanahalli, Bengaluru, Karnataka 560068, India; ORCID: 0000-0002-0658-7581

Clinical Examination

On intra- oral examination, dental caries was present in relation to tooth number 51,52,53,55,61,62,64,65,73,74,84,85,36. Root stumps were present in relation to tooth number 54 and 75. Tooth number 31 was erupting and 41 was missing (Figure 1, 2, 3).

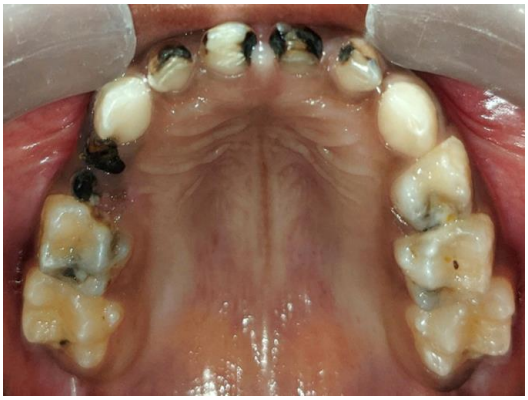

Figure 1: Pre-operative picture of maxilla

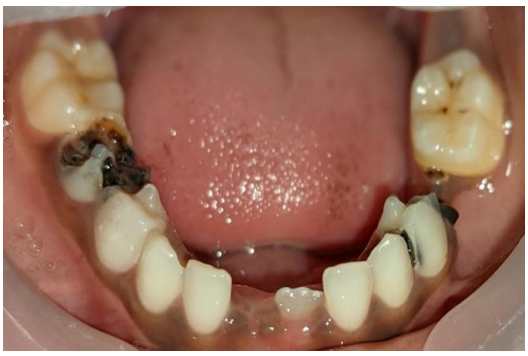

Figure 2: Pre-operative picture of mandible

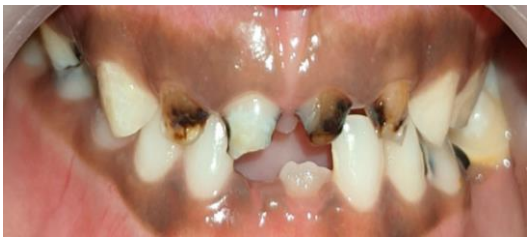

Figure 3: Pre-operative picture in occlusion

Diagnostic Assessment

Orthopantomogram (OPG) was advised. OPG revealed that tooth number 41 was yet to erupt and pulpal involvement of dental caries was present in relation to 52,55,62,64 (Figure 4).

The patient was then referred to a physician for consent to perform extraction and pulp therapy procedures under local anesthesia. The patient was advised chest and abdominal x-ray. Based on the investigations, the diagnosis of situs inversus was confirmed. However, ECG findings were normal and no other abnormality was detected. After obtaining consent from the physician, the patient's dental treatment was initiated (Figure 5).

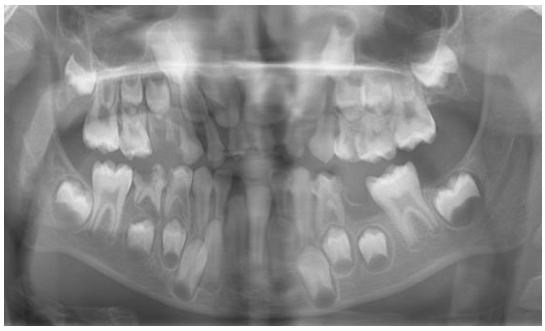

Figure 4: Pre-operative OPG

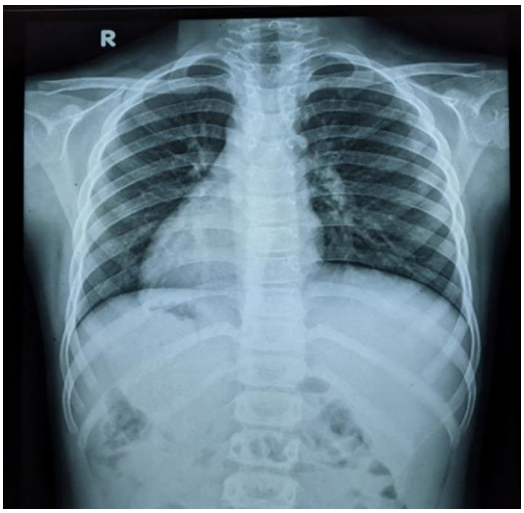

Figure 5: Radiograph of Chest depicting Situs inverus

Therapeutic Interventions

Informed consent form was signed by the parent and the patient before the treatment. Based on the OPG, extraction of 51,54,61,75,84,85 was performed. Pulpectomy in relation to 52,62,64 and pulpotomy in relation to 55 was performed. GIC restoration in relation to 53,65,73,74,36 was performed. Oral prophylaxis was performed. Pit and fissure sealants in relation to 16,26, 46, and fluoride varnish were also applied. A bilateral removable functional space maintainer was given for both the maxillary and mandibular arches (Figure 6, 7, 8, 9).

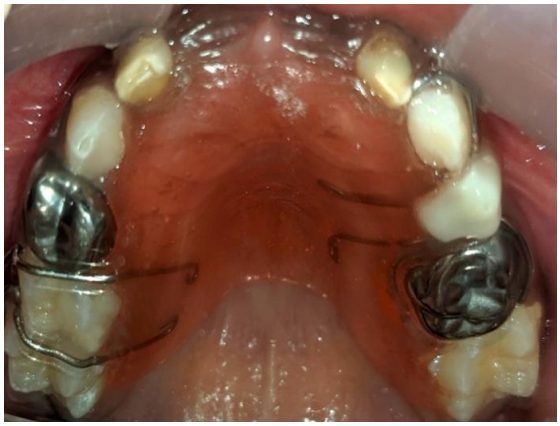

Figure 6: Post-operative picture of maxilla

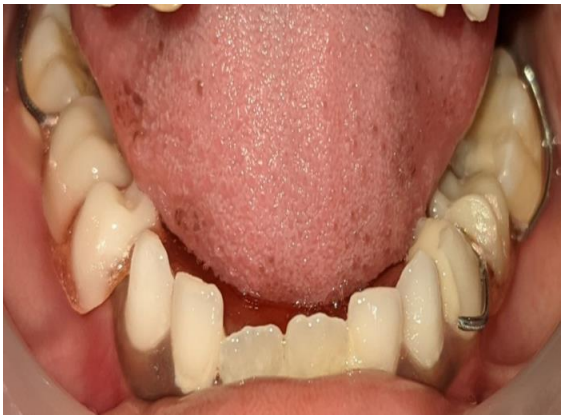

Figure 7: Post-operative picture of mandible

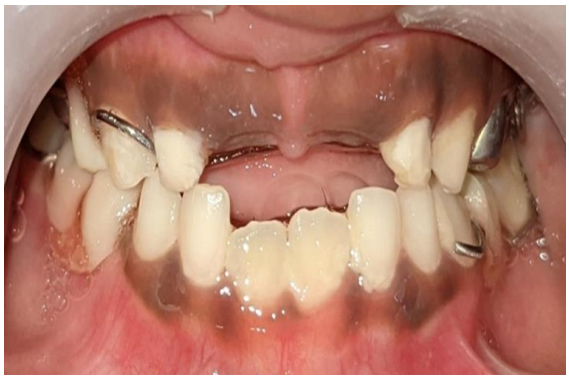

Figure 8: Post-operative picture in occlusion

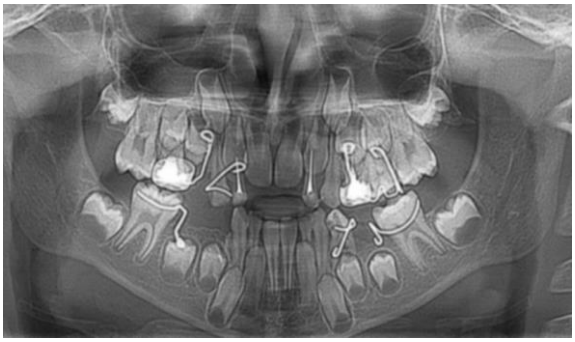

Figure 9: Post-operative OPG

Follow Up

Subsequent visits were performed at 1 day, 1 week, 1 month, and 3 months.

DISCUSSION

In normal human development, the heart lies on the left and the liver on right. The chest and abdominal organs have an asymmetrical arrangement. In Situs inversus, the position of the internal organs is inverted. Situs inversus is short for "Situs inversus viscerum," a Latin phrase meaning "inverted position of the internal organs" [2].

Situs inversus totalisis the term used to describe Situs inversus with dextrocardia. Mirror imaging of abdominal and thoracic viscera is seen. The apex of the heart is located on the right side instead of the left. The right atrium is on the left and vice versa. The normal pulmonary anatomy is also reversed. The stomach and spleen are located on the right and the liver and gallbladder on the left side of the abdomen. The intestines and blood vessels, nerves, and lymphatics are also transposed [2].

Situs inversus was first detected in animals by Aristotle. He considered the condition a visitation from the gods. In 1600, the condition was seen in cadavers [2]. Dextrocardia was first seen by Leonardo da Vinci in 1452-1519. Later, it was recognized by Marco Aurelio Severine in 1643. In 1788, Matthew Baillie described the complete mirror-image reversal of the thoracic and abdominal organs in situs inversus [7, 8].

Situs inversus is believed to be an autosomal recessive genetic condition and an X-linked condition [9]. In Situs inversus totalisis (SIT) there is an absence of a single protein, due to mutation on murine chromosome 12 [10]. The exact cause is unknown, but it as been linked with several other factors such as an autosomal recessive gene with incomplete penetrance, maternal diabetes, cocaine use, and conjoined twinning [11-13].

Situs inversus is present in 0.01% population [7]. Situs inversus is reported to occur in 1 in 8000 to 1 in 25,000 patients as a rare congenital anomaly [14]. No racial predilection exists. The male-to-female incidence is 1:1 [6].

Generally, the condition is asymptomatic unless not associated with congenital heart defects. Great vessels transposition is seen in 5 to 10% of cases and is the most commonly occurring defect [15]. In addition, dextrocardia with a normal abdominal situs has a higher incidence (90-95%) of associated congenital cardiac anomalies including transposition of the great vessels, Atrial septal defects (ASDs), and ventricular septal defect (VSDs) [6]. Comparatively, dextrocardia with situs inversus is associated with a lower incidence (0 to 10%) of congenital heart disease. Other associated congenital anomalies may include duodenal atresia, asplenism, multiple spleens, ectopic kidney, horseshoe kidney, and various pulmonary and vascular abnormalities [4].

The life expectancy of individuals with situs inversus without other congenital anomalies is normal. The risk of acquiring the disease is similar to the same age and sex of other persons. Life expectancy is reduced based on the severity of the defect with cardiac anomalies [6, 16]. However, to prevent mishaps during surgery, recognition of situs inversus is important [4].

It can be diagnosed by chest radiography, electrocardiogram, echocardiography, computed tomography, magnetic resonance imaging, and abdominal ultrasonography. Chest radiography, computed tomography and magnetic resonance imaging

depict the position of the organs. Electrocardiogram, echocardiography, and abdominal ultrasonography provide insight into the functioning of the organs.

In our patient, situs inversus was associated with dextrocardia. The diagnosis was confirmed with chest radiography. No cardiac anomalies were identified on echocardiography. In our literature search, few cases of situs inversus associated with aglossia were found [3, 17, 18].

CONCLUSION

In previous literature, hypoglossia has been reported as an isolated defect associated with situs inversus totalisis. In our patient no other abnormalities were found. To our knowledge, this is the first report of full mouth rehabilitation in a case of situs inversus with no other abnormality. Situs inversus patient will have a normal life span but will need to inform their clinicians of their anatomical mirroring to prevent complications during any medical interventions.

Conflict of Interest: The authors declare that they have no conflict of interest.

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